Clinical Case Report

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Ulcerative colitis with acute pleurisy

A case report and review of the literature

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Abstract

Rationale: Inflammatory bowel diseases (IBD), including ulcerative colitis (UC) and Crohn's disease, are associated with a large number of extraintestinal manifestations. Pulmonary manifestations are infrequently seen in patients with IBD. Moreover, serositis including pleural and pericardial manifestations in UC is rare.

Patient concerns: We report a case of UC with acute pleurisy in a 43-year-old man; review literature; and discuss the diagnosis, differential diagnosis, and treatment.

Diagnoses: Active duodenal ulcer was found using gastroscopy. Multiple ulcers in segmented pattern were noticed in the left hemi-colon using colonoscopy. An UC in active stage was confirmed subsequently by histology.

Intervention: The patient was treated with bifidobacterium tetravaccine tablets, oral mesalazine and mesalazine enemas. The omeprazole and mucosal protective agents were given to treat the duodenal ulcer.

Outcomes: As follow-up, the therapy including oral mesalazine and infliximab regularly was continued and the patient condition was stabilized.

Main lesson: Pulmonary involvement should be considered in patients who develop pleurisy in UC. Infliximab is considered the better available treatment for patients presenting with pleurisy in UC.

Abbreviations: ANA = antinuclear antibodies, ANCA = antineutrophil cytoplasmic antibodies, CD = Crohn's disease, CRP = C-reactive protein, CT = computed tomography, EIM = extraintestinal manifestations, ESR = erythrocyte sedimentation rate, GI = gastrointestinal, IBD = inflammatory bowel diseases, UC = ulcerative colitis.

Keywords: infliximab, pleurisy, ulcerative colitis

1. Introduction

Inflammatory bowel diseases (IBD), including ulcerative colitis (UC) and Crohn's disease (CD), are disorders of chronic inflammation of the gastrointestinal (GI) tract.[1] IBD is associated with a large numbers of extraintestinal manifestations (EIMs). EIMs are present in 10.4% of patients with UC.[2] Common EIMs include arthropathies, mucocutaneous, and ophthalmological manifestations, as well as conditions affecting the hepatobiliary systems. Lung involvement in patients with IBD was firstly reported in 1976.[3] Since then, the respiratory system involved in IBD was increasingly reported in both screening studies and cumulative volume of case reports about.[4,5] Although respiratory changes including upper and small airway diseases, pulmonary vascular diseases, serositis, and other lung diseases can develop at any time in the course of IBD,[6,7] respiratory manifestations seen in patients with IBD remain uncommon.[8] A number of reports had demonstrated that pleural and pericardial manifestations of IBD are rare.[4,9,10] One case of UC with acute pleurisy was reported with review of literatures.

2. Case presentation

The patient provided informed consent for the publication of his clinical and radiological data. The study was approved by the Institutional Ethics Committee of First Affiliated Hospital of Dalian Medical University (Dalian, China).

A 43-year-old man was admitted to the hospital in July 2016 with chronic diarrhea, bloody purulent stool abdominal pain, and fever (up to 39.3°C). The signs included sour regurgitation, heartburn, nausea, anorexia, fatigue, and weight loss of 5 kg within 2 months. There was a history of upper GI hemorrhage from duodenal ulcer 20 years ago. After treatment, vital signs, including cardiac and pulmonary examinations, were shown as normal. Physical examination revealed tenderness at the site of

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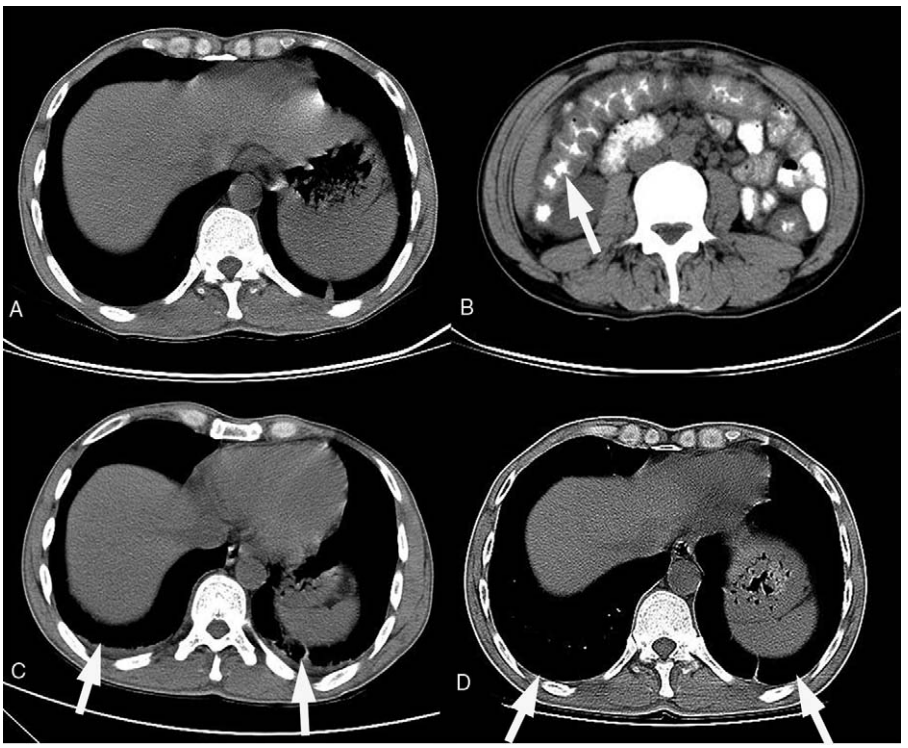

Figure 1. Thorax and abdomen CT images. (A) Image of lung before treatment was normal. (B) Mild dilatation and wall thickening of terminal ileum is shown in abdomen image (arrow). (C) 2-sided basal pleural effusion is shown in lung image (arrows). (D) Pleural effusion was absorbed (arrows).

left lower quadrant of abdomen, without rebound tenderness or guarding. The bowel sounds were active.

For laboratory results, viral hepatitis (A–E) and HIV were shown as negative. The functions of liver and renal were normal. Tumor markers were shown as negative. In stool sample, red blood cells, white blood cells, and pus cells were full of sight under high-power lens. Occult blood was positive. Repeated (3 times) stool cultures for the pathogens were negative. Hemoglobin was 10.6 g/dL and white blood cell count was normal. Albumin was 33.2 g/L. An erythrocyte sedimentation rate of 45 mm/h (normal range, 0 to 20 mm/h) and a C-reactive protein level of 38.2 mg/L (normal range, 0 to 20 mg/L) were found. The perinuclear staining patterns of antineutrophil cytoplasmic antibodies and antinuclear antibodies were positive.

Active duodenal ulcer was found using gastroscopy. Multiple ulcers in segmented pattern were noted in the left hemi-colon using colonoscopy. An UC in active stage was confirmed subsequently by histology. It was normal in computed tomography (CT) scan of lung (Fig. 1A). Mild intestinal dilatation and wall thickening of terminal ileum were found in abdominal image (Fig. 1B).

The patient was treated with bifidobacterium tetravaccine tablets according to previous reports,[11,12] oral mesalazine (4 g/day), and mesalazine enemas (once a day) afterwards. The omeprazole and mucosal protective agents were given to treat the duodenal ulcer. However, the symptoms including diarrhea, bloody purulent stool, abdominal pain, and fever did not resolve after 2 weeks. Meanwhile, the patient experienced obvious chest pain and dyspnea, worsening upon a deep breath. No cough, sputum, and wheezing was shown. There was no history of smoking, lung diseases, allergy, concomitant medications, occupational exposures, pets culturing, or recent travels experience. On physical examination, a pleural friction rub was detected at the base of left lung. No cyanosis, edema, or digital clubbing was found. D-Dimer and cardiac markers were shown as normal in blood gas analysis. Two-sided basal pleural effusion (Fig. 1C) was found in CT images after 2-week treatment, and pleuritis with pleural effusion was confirmed. Steroid treatment could not be chosen as an optimal method due to his active duodenal ulcer. As a result, infliximab (200 mg 1 time) was prescribed. One day later, the symptoms including chest pain, fever, and abdominal pain disappeared. The pleural effusion resolved (Fig. 1D) 3 days later after treatment of infliximab.

As follow-up, the therapy including oral mesalazine and infliximab regularly was continued and the patient condition was stabilized.

3. Discussion

For this active UC patient, lung diseases or heart disease was not found accompanying with UC during the time of chest pain happening. Pulmonary embolism and acute myocardial infarction were also excluded. No evidence was there to support the diagnosis of pneumonia and tuberculous serositis. However, bilateral basal pleural thickening and pleural effusion were found in CT images.

Previous reports showed that sulfasalazine or mesalazine could induce pulmonary disease,[7,13–15] and mesalazine-induced pleural effusion was always associated with pericarditis or pneumonia.[16,17] It was reported that most reactions related to sulfasalazine and mesalazine were seen between 2 and 6 months after commencement of drug administration.[18] It is difficult to differentiate pulmonary disease caused by adverse drug reaction from EIMs in some cases of IBD. The prompt occurrence of

symptoms after drug assumption and the rapid improvement after drug withdrawal demonstrated that pulmonary disease was the adverse drug reaction.[7] However, physicians should be aware that the termination of drugs usage can result in the risk of bowel disease recurrence. This patient's symptoms were evidentially improved, the pleural effusion resolved, and pleural thickening regressed after the use of infliximab. As a result, pleuritis was UC-related and was not induced by mesalazine.

The similarity of embryonic origin between GI and respiratory system provides pathophysiologic basis for respiratory involvement in IBD. First, the colonic and respiratory epithelial cells in both goblet cells and submucosal glands are derived from the same original source of the embryonic foregut. In addition, lymphoid tissue in the submucosa of both lung and GI tract play a critical role in defense of the host mucosa.[4,6,19,20] Acute pleurisy was considered an abnormal immune response with UC.

The treatment of IBD-related respiratory involvement is associated with the specific pattern of involvement.[8] Neither colectomy nor classic nonsteroidal modifying drugs including immunosuppressants have demonstrated any effect in control of the respiratory manifestations of IBD. Most cases of IBD, necrotic nodules, and serositis are very sensitive to steroid drugs. Steroid drugs are effective and for the long-term control of large airways IBD-related disease.[21] Infliximab is helpful for rapid improvement of the lung and skin lesions in a patient with severe UC.[22] For this patient, GI hemorrhage has occurred due to peptic ulcer before, and the ulcer disappeared with the treatment of antacids. At that time, the typical duodenal ulcer was single in the bulb shown as a feather in endoscopy together with Hp positive. After PPI treatment for 1 month, the ulcer was healed. Therefore, the duodenal ulcers were not considered as a part of UC. The active duodenal ulcer is happening during the time of pleurisy, consequently steroids should be avoided although it may be the first choice of the treatment for the occurrence of pleurisy in this UC patient. Therefore, infliximab was utilized and proved effective for the remarkable remission of pleurisy and UC.

In conclusion, pulmonary involvement should be considered when pleurisy developed in UC. Early diagnosis and effective treatment are important for these patients. Infliximab is considered a better treatment method for patients presenting with pleurisy in UC.

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